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Brief Report: Self-Injurious Behaviors in Preschool Children with Autism Spectrum Disorder Compared to Other Developmental Delays and Disorders

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Abstract

We compared the prevalence of self-injurious behaviors (SIB) in preschoolers aged 30–68 months with autism spectrum disorder (ASD) (n = 691) versus other developmental delays and disorders (DD) (n = 977) accounting for sociodemographic, cognitive, and medical factors. SIB prevalence was higher in ASD versus all DD [adjusted odds-ratio (aOR) 2.13 (95% confidence interval (95% CI) 1.53, 2.97)]. In subgroup analyses, SIB prevalence was higher in ASD versus DD without

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Author Contributions GNS conceptualized the initial proposal, cleaned and analyzed the data, wrote the first draft of the manuscript. CD, SAR and CRR, provided feed-back on the initial proposal, contributed in the interpretation of the findings and in the writing of the different versions of the manuscript. LL and RAV contributed to the interpretation of the findings and the writing of the different versions of the manuscript.

Disclaimer The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

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Compliance with Ethical Standards

Conflict of interest All authors do not have any conflict of interest to disclose.

Ethical Approval All procedures performed in studies involving human participants were in accordance with ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent All persons who participated in SEED provided written informed consent.

ASD symptoms [aOR 4.42 (95% CI 2.66, 7.33)], but was similar between ASD and DD with ASD symptoms [aOR 1.09 (95% CI 0.68, 1.77)]. We confirmed higher prevalence of SIB in ASD versus DD, independent of confounders. In children with DD, SIB prevalence increased with more ASD symptoms. These findings are informative to clinicians, researchers, and policymakers.

Keywords

Self-injurious behaviors; Prevalence; Autism spectrum disorder; Autism; Developmental delays and disorders

Introduction

Self-injurious behaviors (SIB) are a group of self-inflicted, harmful behaviors, such as arm biting and head banging, that may result in injuries to self (Dempsey et al. 2016; Fee and Matson 1992) and increase the costs of care (Ianuzzi et al. 2015; Minshawi et al. 2014). The etiology of SIB is complex, involving biological and non-biological factors. Further, research has demonstrated that SIB can acquire an operant function within a child's environment, including socially mediated functions such as escaping from demands or obtaining attention and non-socially mediated functions such as self-regulation of arousal (Carr 1977; Guess and Carr 1991; Iwata et al. 1994; Kurtz et al. 2003).

SIB have been documented in individuals with developmental delays and disorders (DD), including autism spectrum disorder (ASD). The reported prevalence of SIB in individuals with DD varies between 4 and 32% (McClintock et al. 2003; Ruddick et al. 2015; Schroeder et al. 2014). In individuals with ASD, the reported SIB prevalence ranges between 20 and 53% (Baghdadli et al. 2003; Duerden et al. 2012; Soke et al. 2016). Further, studies have found that the presence of a co-occurring ASD diagnosis in those with DD increased the risk of SIB (Cervantes and Matson 2015; MacLean and Dornbush 2012; MacLean et al. 2010; Matson and Rivet 2008; McClintock et al. 2003). Studies have compared the prevalence of SIB between those with ASD and DD and reported higher prevalence of SIB in the former group (Bodfish et al. 2000; Dominick et al. 2007; Matson and Rivet 2008; Richards et al. 2012; Schroeder et al. 2014; Wiggins et al. 2015b). For example, Schroeder et al. (2014) found that the mean score of SIB was higher in children with ASD or at risk for ASD versus those with Down syndrome (DS) or with atypical development. Similarly, Richards et al. (2012) found higher SIB prevalence in those with ASD versus DS. However, no major difference was noted between those with ASD and fragile X syndrome.

Although the above studies documented higher prevalence of SIB in individuals with ASD versus DD, it is unclear whether this finding could be due to confounding factors. Differences in child and maternal characteristics [e.g., child sex, intelligence quotient (IQ) and adaptive scores, gastrointestinal and sensory problems; maternal education] have been documented between those with ASD and DD (DiGuiseppi et al. 2016; Dominick et al. 2007; Valicenti-McDermott 2006; Ventola et al. 2007; Wiggins et al. 2015b). Further, some of these characteristics are associated with SIB (Baghdadli et al. 2003; Duerden et al. 2012; Soke et al. 2017). Therefore, comparing SIB prevalence between individuals with ASD and DD can be biased if adjustments are not correctly made for the above characteristics.

Furthermore, most of the above-mentioned studies (e.g., Dominick et al. 2007; MacLean et al. 2010) used clinical samples, which may have included high proportions of severe cases, leading to possible biased estimates of SIB. Lastly, data comparing SIB prevalence between ASD and others DD in preschool children are lacking since most published studies, except for Schroeder et al. (2014), included samples of older children or adults. These data in preschoolers are needed to develop targeted prevention strategies and inform early intervention programs. Therefore, the objective of this study was to compare SIB prevalence between preschoolers with ASD and DD in a large and community-based sample, adjusting for various confounding factors.

Methods

Study Design

We used a case–control study and included data from the phase 1 of the Study to Explore Early Development (SEED). Data collection was approved by the Centers for Disease Control and Prevention and each site's institutional review board.

Participants

Children with a final classification of ASD and DD from six sites in the United States (California, Colorado, Georgia, Maryland, North Carolina, and Pennsylvania) who had data on SIB were included. All six SEED sites followed a common protocol for recruitment and data collection procedures, which are presented in detail in other publications (DiGuiseppi et al. 2016; Schendel et al. 2012; Singer et al. 2016; Wiggins et al. 2015a, b). Briefly, at each site, children aged 30–68 months, with a caregiver fluent in English or, at two sites (California and Colorado), English or Spanish, were enrolled. Potential cases of ASD and DD were identified from special education programs, clinics and other providers of disabilities services. Further, parents of children with an ASD or DD diagnosis obtained from a community provider were allowed to self-refer for enrollment into SEED Phase 1. Data collection in SEED included a caregiver interview, various self-administered questionnaires, and an in-person clinical evaluation (Schendel et al. 2012). An ASD screener, the Social and Communication Questionnaire [SCQ] (Rutter et al. 2003) was administered to the caregivers after enrollment. All children received a general developmental evaluation using the Mullen Scales of Early Learning [MSEL] (Mullen 1995) during a clinic visit. Children who screened positive on the SCQ [i.e., score ≥ 11] (Wiggins et al. 2007), had a previous diagnosis of ASD, or were suspected of possible ASD by a clinician during the clinic visit also received an ASD evaluation, which consisted of the Autism Diagnostic Observation Schedule [ADOS] (Lord et al. 2000a) and the Autism Diagnostic Interview-Revised [ADI-R] (Lord et al. 1994). Caregivers also completed the Vineland Adaptive Behavior Skills [VABS-2] (Sparrow et al. 2005) as part of the ASD evaluation, or if the child had a score < 78 on the MSEL. All cases of ASD met criteria on the Diagnostic and Statistical Manual for Mental Disorders-IV-edition-text revision [DSM-IV-TR] (American Psychiatric Association 2000), and scored above the cut-offs for ASD on the ADOS and the ADI-R based on an algorithm developed by SEED clinicians (Wiggins et al. 2015b). This algorithm considered ASD symptoms relevant to both the 4th and 5th editions of the DSM. Children were classified as DD if they had a DD condition reported by

parents on the caregiver interview, did not show a risk of ASD on the SCQ, or did show a risk of ASD on the SCQ but did not meet the criteria for ASD during the ASD evaluation. The DD group was divided post hoc into two subgroups based on different ASD-specific symptom profiles: DD with ASD symptoms and DD without ASD symptoms. DD with ASD symptoms received a comprehensive developmental evaluation for ASD because of a SCQ score ≥ 11 or a previous ASD diagnosis but did not meet the SEED threshold for ASD criteria. DD without ASD symptoms received a limited developmental evaluation since they did not have a previous ASD diagnosis and scored below 11 points on the SCQ (Wiggins et al. 2015b). The DD group included various conditions such as intellectual disability, language disorder, hearing and vision impairments, and attention-deficit/hyperactivity disorder [ADHD] (Wiggins et al. 2015b).

Measures

The outcome of interest was parent report of SIB based on question #17 of the SCQ, which asks “Does the child ever injure her/himself deliberately, such as by biting her/his arm or banging her/his head (yes/no).” We adjusted for child characteristics (sex, age, IQ scores, gastrointestinal and sleep problems) and family characteristics (mother’s education and race, mother’s age, mother’s psychiatric conditions, family income). Data on these potential confounders were obtained from various caregiver interviews, other instruments (MSEL, Children’s Sleep Habit Questionnaire, Gastrointestinal Symptom Inventory) and birth certificates. The Children’s Sleep Habit Questionnaire [CSHQ] (Owens et al. 2000) is a parent completed questionnaire that assess sleep problems in a child (e.g., difficulties falling and staying asleep, night waking, and daytime sleepiness) during a recent typical week. The CSHQ provides an overall total score and domain specific scores with high scores indicating more sleep problems and has been previously used in preschoolers by other researchers (Goodlin-Jones et al. 2008). The Gastrointestinal Symptom Inventory is a SEED developed parent completed questionnaire that provides data on current gastrointestinal problems in a child (e.g., vomiting, diarrhea, constipation, abdominal pain) that occurs on a regular basis (i.e., > 2 times per month). Maternal physician-diagnosed psychiatric conditions were reported in the maternal medical history form.

Analytic Strategy

We compared characteristics of children with ASD and DD using Chi square tests for categorical variables and *t* tests for continuous variables. To assess for potential confounders, we also evaluated the characteristics associated with SIB versus no SIB, among all study children (i.e., ASD and DD combined) using logistic regression. Any variable associated with both case status and with SIB at the p value ≤ .20 in the univariate analyses was considered as a potential confounder and included in the adjusted analyses. Further, any variable associated with SIB was also included as a precision variable in the adjusted analyses. We compared unadjusted and adjusted odds of SIB between children with ASD and DD using non-linear mixed models including study site as a random effect to account for within-site clustering. As a secondary analysis, we compared the prevalence of SIB between (a) children with ASD and the two DD subgroups (with and without ASD symptoms), and (b) between the two DD subgroups, using the same analytic strategy described above. Since the SCQ was used as a screener for ASD and SIB is one of the items

on the SCQ, some DD children could have met the cut-off and be classified as DD with ASD symptoms because of SIB. Therefore, we repeated all the above comparisons after excluding DD children who had SIB and a SCQ score of 11.

Results

A total of 691 children with ASD and 977 children with DD were included in these analyses. Among the 977 total children in the DD group, 297 (30.4%) were classified as DD with ASD symptoms and 680 (69.6%) were classified as DD without ASD symptoms. The comparison of characteristics between children with ASD and DD is presented in Table 1. We identified differences on a number of child and maternal characteristics between children with ASD and children with DD. SIB were reported in 29.4% of children with ASD and in 12.4% of children with DD (8.5% of DD with ASD symptoms and 3.9% of DD without ASD symptoms). The associations between SIB and different characteristics examined are presented in Table 2. All the characteristics examined were significantly associated with SIB and therefore included in the adjusted analyses. The unadjusted and adjusted odds of SIB between children with ASD versus all DD (i.e., including the entire DD group), and between children with ASD and each of the two subgroups of DD children, are included in Table 3. The unadjusted and adjusted odds of SIB were significantly higher in children with ASD versus all DD children. Similarly, the unadjusted and adjusted odds of SIB were higher in children with ASD versus DD without ASD symptoms, and also in DD children with ASD symptoms versus DD children without ASD symptoms. No significant differences were found between children with ASD and children with DD with ASD symptoms. Only six children (i.e., 0.6% of all DD children and 2% of DD children with ASD symptoms) had SIB and a SCQ total score of 11. Excluding these children did not change our results (data not shown). The comparison of characteristics between the two sub-groups of DD children is presented in a Supplemental Table (Table 1S). The two sub-groups differed in all the characteristics examined except for the presence of maternal depression and anxiety.

Discussion

Using a large, community-based sample of preschoolers, SIB prevalence was higher in those with ASD compared to DD. In a secondary analysis, the prevalence of SIB in children with DD was higher when ASD symptoms were present. To our knowledge, this is the first study to compare the prevalence of SIB between ASD and DD in a large sample of preschoolers.

Similar to other studies (Bodfish et al. 2000; Dominick et al. 2007; McClintock et al. 2003, Wiggins et al. 2015b), we confirmed higher prevalence of SIB in children with ASD versus DD, even after adjusting for various characteristics. Therefore, the higher SIB prevalence in children with ASD may be a valid finding that cannot be completely attributed to confounding factors. We also found that the magnitude of the difference between children with ASD and those with DD increased when the comparison was restricted to DD children without ASD symptoms, and the difference dissipated when children with ASD were compared to DD children with ASD symptoms. Therefore, as reported by others (MacLean and Dornbush 2012; Matson and Rivet 2008; McClintock et al. 2003; Moss et al. 2012; Staley et al. 2008; Wiggins et al. 2015b), we confirmed the association between ASD

symptoms with increased prevalence of SIB in those with DD. Therefore, in line with a study by Richards et al. (2012) in those with fragile X and DS, SIB occurrence appears to be more dependent on the presence of ASD symptomatology—even at a subthreshold level—than on an ASD diagnosis per se. Clinicians should consider assessing the presence of ASD symptoms among children with DD, and when present, consider evaluating the occurrence of SIB. Our findings that SIB was more common in children with ASD versus children with DD (with or without ASD symptoms) were quite similar to those reported by Wiggins et al. (2015b), even though these authors used a different maternal interview instrument to assess SIB in SEED and did not adjust for potential confounders.

It is important to acknowledge that since SIB is one item on the SCQ, this could have influenced SCQ score and move some DD children as DD with ASD symptoms. However, our data did not support this possibility, since this affected only a very small proportion of DD children (0.6%) and excluding them did not affect our findings.

The reasons behind the higher prevalence of SIB in individuals with ASD versus DD is not completely understood and need to be explored. In this study, we adjusted for multiple child and maternal characteristics. Low IQ is a known risk factor for SIB (McClintock et al. 2003), and we similarly found that lower IQ scores were associated with more SIB. We also found that IQ scores were lower in children with ASD versus DD, but the difference in SIB prevalence between the two groups persisted after adjustment for IQ. Thus, our data suggest that IQ scores alone cannot explain the difference in SIB prevalence between the two groups. Therefore, other unaccounted factors in our study, such as deficits in executive functions (EF) and adaptive behaviors skills, or their interactions may be involved in SIB occurrence (Gilotty et al. 2002; McClintock et al. 2003; Oliver and Richards 2015; Richards et al. 2012; Turner 1999). EF are a collection of cognitive abilities required for executing and controlling goal-directed behaviors in a constantly changing environment (Jurado and Rosselli 2007). Deficits in EF (i.e., inability to generate, plan, shift and inhibit behavior) may compromise the ability to control repetitive behaviors (Lopez et al. 2005; Turner 1999). Oliver and Richards (2015) proposed a model that elucidated a potential role of deficits in EF in SIB based on the co-occurrence of SIB, impulsivity, and repetitive behaviors. These authors suggested that all these three behaviors could have the same etiology linked to difficulties in regulating behaviors due to deficits in EF. Differences in the types of deficits in EF have been documented between children with ASD and DD. For example, a recent review by Craig et al. (2016) on deficits in EF in those with ASD and ADHD indicated both similarities and differences. One important difference was the documentation of abnormalities in the area of cognitive flexibility/perseveration that affected mostly those with ASD and may result in inability to shift or disengage from an activity and occurrence of persistent repetitive behaviors, including SIB. Further, Lopez et al. (2005) reported associations between deficits in specific types of EF (cognitive flexibility, working memory, and response inhibition) and the occurrence of restricted, repetitive behaviors in those with ASD. It is possible that the same mechanism that leads to restricted, repetitive behaviors is also involved in the occurrence of SIB as suggested by Oliver and Richards (2015). Thus, there is a need to evaluate the association between deficits in EF and SIB in large samples of children with ASD.

Adaptive behavior skills are age-appropriate skills, such as self-care, expressive and receptive language, toileting, that allow individuals to function independently in a society (Kanne et al. 2011).

In general, greater deficits in adaptive behavior skills have been documented in individuals with ASD versus DD (Matson et al. 2009; Perry et al. 2009; Rodrigue et al. 1991). It has also been reported that in individuals with intellectual disability, a comorbid ASD diagnosis is associated with more deficits in adaptive behavior skills (Matson et al. 2009). Since low adaptive behavior skills have been associated with SIB in those with ASD (Baghdadli et al. 2003; Duerden et al. 2012; Soke et al. 2017), it is possible that the greater deficits in adaptive behaviors skills in individuals with ASD may contribute to the higher prevalence of SIB in ASD compared to those with DD. However, we were unable to examine the effects of adaptive behavior skills since data were only available for a small group of DD children.

The strengths of this study include (1) its large community sample of children with well-characterized diagnoses of ASD and DD, (2) the younger age of the sample compared to past studies, (3) documentation of SIB using a standardized instrument; (4) adjustments for various factors, and (4) comparisons of subgroups of DD children. However, some limitations should be noted. We included data from six study sites in the United States, which do not constitute a representative sample of children with ASD and DD. Therefore, these findings may have limited external validity. Data on other variables, such as adaptive behavior skills and sensory processing abnormalities, which are known risk factors for SIB, were not available in most DD children, and therefore the contribution of these factors could not be assessed. We did not have data on the type and amount of interventions received by the participants, as this may also influence the proportion of children with SIB. Lastly, since we did not have information on the severity of SIB and parental report of SIB was not confirmed by clinicians, it is possible that parents may have reported less severe and transient SIB, which can be present even in typically-developing children (Berkson and Tupa 2000; Symons et al. 2005). Therefore, it is possible that we may have overestimated the prevalence of SIB in this study in both ASD and DD groups. However, the prevalences in our study were within the ranges of those reported in past studies.

In summary, this study found a higher prevalence of SIB in preschool children with ASD versus DD, after accounting for various other factors, thus validating findings from other studies. Further, we also confirmed the relationship between ASD symptoms and increased prevalence of SIB in the DD group. These findings have clinical, research, and policy implications. Because of the younger age of participants in this study compared to others, these data can inform clinicians working with young children with ASD and DD and suggest the need to screen for SIB. Further, researchers could use our findings to develop targeted prevention strategies, inform early intervention programs, and design future studies that will assess the potential reasons explaining the higher prevalence of SIB when ASD symptoms are present. Such studies could provide a better understanding of SIB, which can inform interventions. Policymakers may use these findings for allocation of resources for future research and service planning to provide timely intervention for SIB and mitigate its consequences. Future studies should consider including details on the severity and types of SIB, since others have found differences in the types of SIB between those with ASD and

DD (Buono et al. 2010; Rice et al. 2016; Richards et al. 2012). Future studies should also consider other factors, such as genetic conditions, sensory abnormalities, adaptive behavior skills, type and duration of interventions received .

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table 1

Comparisons of child and family characteristics between preschool children with ASD and DD in the SEED

Variable	Autism spectrum disorder (n = 691)	Developmental delays and disorders (DD) (n = 977)	p Value[§]
Child sex: n (%)			< **.001**
Female	125 (18.09)	328 (33.57)	
Male	566 (81.91)	649 (66.43)	
Child age (months)			0.65
Mean (SD)	55.70 (6.83)	55.86 (7.20)	
Child intelligence quotient scores			< **.001**
Mean (SD)	66.87 (20.00)	87.75 (21.02)	
Child gastrointestinal problems: n (%)			< **.001**
Yes	222 (32.13)	200 (20.47)	
No	421 (60.93)	716 (73.29)	
Missing	48 (6.94)	61 (6.24)	
Child sleep scores			< **.001**
Mean (SD)	47.98 (9.99)	45.58 (8.87)	
Maternal age (years)			0.94
Mean (SD)	31.63 (5.54)	31.62 (5.74)	
Maternal race: n (%)			0.27
African American	139 (20.12)	182 (18.63)	
Other	120 (17.37)	148 (15.15)	
White	432 (62.51)	647 (66.22)	
Maternal education: n (%)			0.33
No college degree	329 (47.61)	444 (45.45)	
College degree or higher	361 (52.24)	533 (54.55)	
Missing	1 (0.15)	0	
Maternal depression: n (%)			**0.17**
Yes	179 (25.90)	225 (23.03)	
No	512 (74.10)	752 (76.97)	
Maternal anxiety disorder: n (%)			**0.009**
Yes	66 (9.55)	60 (6.14)	
No	625 (90.45)	917 (93.86)	
Maternal intellectual disability: n (%)			**0.06**
Yes	43 (6.22)	41 (4.20)	
No	648 (93.78)	936 (95.80)	
Family income: n (%)			0.54
$10,000 to < 30,000	162 (23.44)	211 (21.60)	
$30,000 to < 70,000	175 (25.33)	253 (25.90)	
$70,000 to < 110,000	163 (23.60)	239 (24.46)	
$> 110,000	163 (23.60)	219 (22.42)	
Missing	28 (4.03)	55 (5.62)	

SD standard deviation

[§] Bold indicates significant p values (≤ .20)

Table 2

Unadjusted associations between SIB and different covariates in preschool children with ASD and DD in the SEED

Variable	Odds ratio and 95% confidence interval[§]	p Value
Child sex		**0.07**
Female	**0.77 (0.58, 1.02)**	
Male (Ref.)	1.00	
Child age (1 month)	**0.99 (0.97, 1.01)**	**0.16**
Child Intelligence quotient scores (1 unit)	**0.98 (0.97, 0.98)**	< **.001**
Child gastrointestinal problems		**0.004**
Yes	**1.63 (1.24, 2.14)**	
No (Ref.)	1.00	
Child sleep scores (1 unit)	**1.06 (1.05, 1.08)**	< **.001**
Maternal age (1 year)	**0.93 (0.91, 0.95)**	< **.001**
Maternal race/ethnicity		**0.002**
African American	1.31 (0.98, 1.76)	
Other	**0.59 (0.40, 0.89)**	
White (Ref.)	1.00	
Maternal education		< **.001**
No college degree	**2.92 (2.26, 3.77)**	
College degree or higher (Ref.)	1.00	
Maternal depression		**0.002**
Yes	**1.53 (1.17, 2.00)**	
No (Ref.)	1.00	
Maternal anxiety disorder		**0.03**
Yes	**1.60 (1.05, 2.41)**	
No (Ref.)	1.00	
Maternal intellectual disability		< **.001**
Yes	**2.88 (1.82, 4.54)**	
No (Ref.)	1.00	
Family income		< **.001**
$10,000 to < 30,000	**4.09 (2.82, 5.95)**	
$30,000 to < 70,000	**1.70 (1.15, 2.52)**	
$70,000 to < 110,000	1.01 (0.66, 1.56)	
$> 110,000 (Ref.)	1.00	

[§]Bold indicates significant estimates (p value ≤ .20)

Table 3

Unadjusted and adjusted odds of SIB in preschool children with ASD compared to DD in the SEED

Groups compared	Unadjusted odds ratio and 95% confidence interval*	Adjusted odds ratio§ and 95% confidence interval*
Autism spectrum disorder versus all developmental delays and disorders	**2.94 (2.29, 3.78)**	**2.13 (1.53, 2.97)**
Autism spectrum disorder versus developmental delays and disorders with ASD symptoms	1.07 (0.75, 1.53)	1.09 (0.68, 1.78)
Autism spectrum disorder versus developmental delays and disorders without ASD symptoms	**7.03 (4.76, 10.37)**	**4.42 (2.66, 7.33)**
Developmental delays and disorders with ASD symptoms versus developmental delays and disorders without ASD symptoms	**6.55 (4.33, 9.92)**	**4.09 (2.44, 6.86)**

*Bold values indicate significant estimates (p value < .05)

§Theses analyses were adjusted for child sex, age, intelligence quotient, sleep scores, and presence of gastrointestinal problems; maternal age, education and race, depression, anxiety disorder, and intellectual disability; and family income